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Case Report

Treatment of large ameloblastic fibroma: a case report

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Abstract: Ameloblastic fibroma (AF) is an extremely rare true mixed benign tumor that can occur in either the mandible or the maxilla, but is most frequently found in the posterior region of the mandible. It usually occurs in the first two decades of life and is associated with tooth enclosure, causing a delay in eruption or altering the dental eruption sequence. AF is diagnosed on routine radiographic evaluation and is clinically and radiographically similar to ameloblastic fibrodontoma and odontoma, which makes an accurate diagnosis mandatory. There is controversy in the literature as to whether treatment should be conservative or agressive. A conservative treatment strategy, such as enucleation and curettage, is usually sufficient. However, extensive lesions require radical treatment. We describe a case of ameloblastic fibroma with a very unusual clinical manifestation: it demonstrated considerable extension but no associated impacted tooth, was located in the anterior region of the mandible, and became symptomatic in the fifth decade of life. A radical surgical approach was taken, with immediate reconstruction. (J Oral Sci 51, 293-296, 2009)

Keywords: ameloblastic fibroma; odontogenic tumors; epithelium; ectomesenchyme.

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Introduction

Ameloblastic fibroma (AF) is a rare mixed odontogenic tumor that usually occurs in young patients (1), being diagnosed at a mean age of 15 years. It can appear in either the maxilla or mandible, with the posterior region of the mandible as its most common anatomic site (2). Edema and/or an increase in volume are the main signs of AF, although most cases are asymptomatic. Diagnosis is generally made through routine radiographic examinations performed to look for an impacted tooth as a cause of the swelling (3). Primary lesions can usually be sufficiently treated conservatively, with enucleation or curettage, and the tumor has a low rate of recurrence (4).

We describe a case of ameloblastic fibroma that differed from the standard clinical manifestation of the disease. We discuss features such as age at presentation, anatomic location, tooth enclosure, and extension.

Case Report

A 45-year-old man presented with a 4-year history of intraoral swelling, accompanied by difficulty in swallowing and speech. He reported having no pre-existing medical conditions, fever, or other signs of infection. A physical examination revealed facial asymmetry, with an increase in volume extending from the right symphysis to the body of the mandible (14 cm) (Fig. 1a). There was a semi-solid, non-tender intraoral tumor, with poorly defined boundaries and a smooth surface. This lesion was associated with a compromised vestibular fornix and severe tooth mobility (Fig. 1b). Radiographic examinations revealed a multilocular lesion, root resorption of the anterior teeth, and cortical bone expansion in the anterior-posterior and

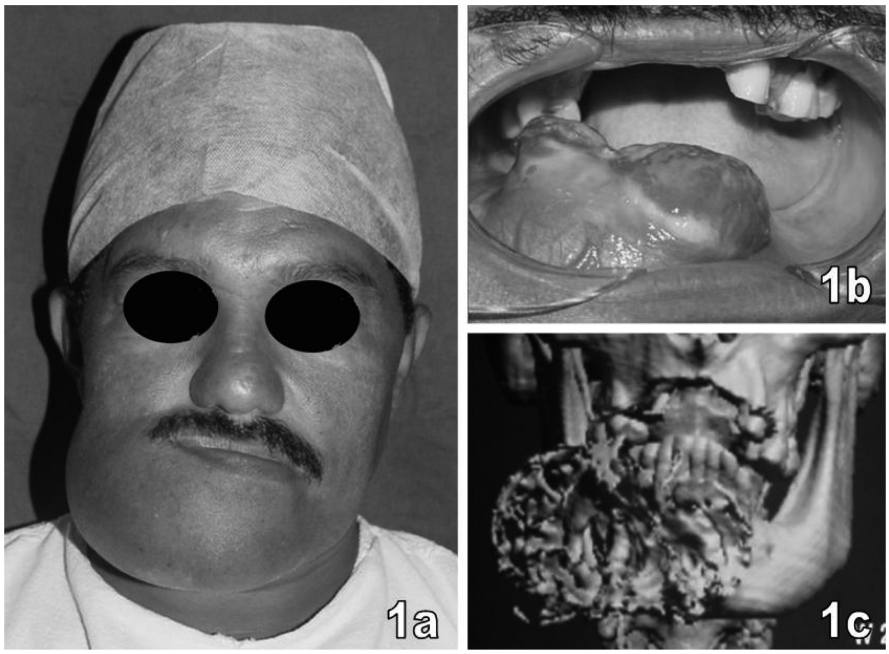

Fig. 1 (a) Frontal view showing extensive increase in volume on the right side of the face; (b) Intraoral view of tumor mass; (c) 3D tomographic reconstruction, showing cortical expansion and fenestration.

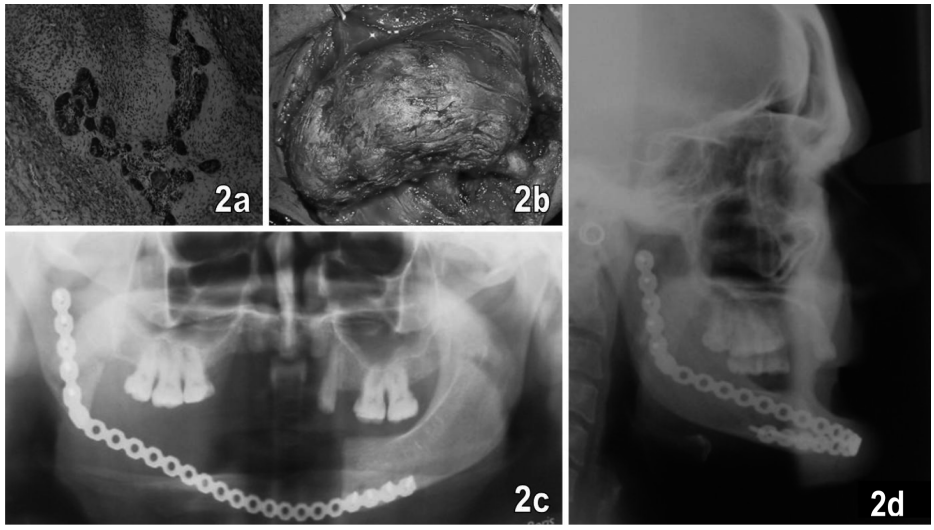

Fig. 2 (a) Histopathologic examination: long, narrow cords of odontogenic epithelium in a richly cellular, primitive mesenchymal stroma (H-E staining, ×100); (b) Surgical exposure of the lesion; (c, d) Orthopantographic examination and cephalometric profile at seven years' follow-up.

superior-inferior planes. Tomographic examination through 3D reconstruction indicated vestibular fenestration of the cortical bone, with involvement of lingual cortical bone as the lesion extended to the posterior region (Fig. 1c). As aspiration was negative, incisional biopsy was performed. Macroscopically, whitish soft tissue of rubbery texture was observed. The microscopic examination revealed a proliferation of odontogenic epithelial tissue and highly cellular mesenchymal tissue. Microscopically, the tumor was composed of cell-rich mesenchymal tissue resembling the primitive dental papilla admixed with proliferating odontogenic epithelium (Fig. 2a). Based on the clinical,

imaging, and microscopic data, a diagnosis of AF was established. Partial mandibular resection was performed, with immediate reconstruction by rigid fixation (2.4 mm) (Fig. 2b). The patient has been followed up for 7 years; the outcome has been functionally and aesthetically satisfactory (Figs. 2c, d) and there have been no signs of recurrence.

Discussion

AF is usually diagnosed between the first and second decades of life, in contrast to the present case involving an adult patient in the fifth decade of life. It is hard to say that delay in seeking treatment was the sole reason for the late diagnosis as the patient had had symptoms for only 4 years and it would be difficult to prove the lesion was present but asymptomatic for over 30 years until the late diagnosis in this case.

This condition is generally associated with enclosed teeth in the posterior region of the mandible, angle, and/or ramus. The radiographic appearance may vary from a small unilocular lesion to an extensive multilocular lesion (5). Bone expansion and tooth dislocation are common findings. In the case reported, radiographic examinations revealed root resorption and no enclosed teeth; these are unusual findings for this kind of lesion. The tomography images revealed fenestration of the cortical bone, a characteristic of extensive and long-standing lesions but rarely observed in AF.

Histologically, AF is a true mixed tumor, as it has both mesenchymal and epithelial neoplasic components with no associated calcified tissue (2). Ameloblastic fibro-odontoma (AFO) is defined as a tumor that shares many features with AF but has enamel and dentin in its interior (1). Some authors consider this lesion an intermediate stage in the development of an odontoma, with the primary stage of formation being AF (6). As some odontomas have similar histological features to AF and AFO, clinical findings are fundamental to the differentiation of these three pathological entities. If all cases occurred following the particular developmental stage outlined above, AF would affect young patients, odontoma would occur in elderly patients, and AFO would be seen in an intermediate age group. However, this is not the case. In the past, AF was regarded as a variant of ameloblastoma, but it has recently been considered to be of odontogenic epithelial origin.

The treatment performed in the present case differs from that in standard AF cases due to the aggressiveness of the lesion, with its considerable extension and soft tissue involvement. Mandibular resection was performed, followed by immediate reconstruction. The fixation model was constructed on a dry mandible, which is a less burdensome option in comparison to the stereolithographic model. This proved to be a fast, simple method, suitable for use when there is a lack of resources.

There is disagreement regarding forms of treatment due to the different recurrence rates described in the literature. Trodahl (3) and Chen et al. (6) reported high recurrence rates (43.5 and 91.5%, respectively), stating that many tumors were primary, resulting from unsatisfactory enucleation rather than being recurrent. Another factor is that recurrent cases require a more radical surgical procedure. Zallen et al. (7) recommend block resection as the initial treatment for AF, due to the high rate of recurrence in cases in which a conservative approach was initially performed. However, Dallera et al. (8) presented a long-term follow-up study of six cases and found that good results were obtained in cases in which a conservative approach was performed with enucleation and curettage. The reason for the discrepancies in recurrence rates described in the first three studies and those obtained in the last mentioned study is uncertain and suggests that the cause of recurrence is incomplete removal and presence of satellite tumors at the edge of the lesion.

The prognosis for lesions of this proportion is dubious. In young patients in whom resection will result in mutilation and in small lesions that respond well to conservative treatment, we recommend enucleation and curettage. However, in extensive lesions and the involvement of adjacent tissues, as in the case described here, we recommend block resection.

Regardless of the form of treatment, patients with AF must be followed up for a long period to enable the early detection of possible recurrence or development of ameloblastic fibrosarcoma, which is the malignant counterpart of AF (9). In the case described here, the patient has been followed up for seven years with no signs of recurrence or sarcomatous transformation, thereby demonstrating the success of the treatment.

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